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CLINICAL COMMENTARY

Cholesteatoma of the frontal sinus

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KEYWORDS
Cholesteatoma;
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Summary
Purpose of study: Cholesteatoma of the paranasal sinuses is a rare pathology. A review of the literature reported less than 30 cases. These lesions mainly involve the frontal sinus.
Case report: A 25-year-old man presented with a right painful fronto-orbital mass associated with an ipsilateral eyelid oedema and a fever as high as 40 °C. He experienced a general epileptic seizure requiring his admission in an intensive care unit.

CT-Scan with iodine injection evidenced the opacification of the right frontal sinus with bone lysis of the posterior wall. Complete surgical resection of a cystic structure containing keratin material was performed via eyebrow incision. The pathological examination confirmed the diagnosis of sinus cholesteatoma. Neurological signs entirely disappeared after surgery. Craniofacial MRI realized 2 months later showed no sign of recurrence. Obliteration of the right frontal sinus was performed 4 months later.
Conclusion: Although benign, cholesteatoma can spread to the surrounding structures leading to several complications including infections that can be life-threatening for the patient. CT-scan and MRI are useful examinations for diagnosis and follow-up. Complete surgical resection is required in order to avoid recurrence.

Introduction

Cholesteatoma is generally a pathology of the middle ear, yet it can exceptionally occur in the frontal sinus. A review of the literature in English reported less than 30 cases [1,2,3].

Facial deformities, ptosis, visual impairment are the presenting signs of this pathology [4,5]. Cases of malignant degeneration and death were even reported [3,6].

We report the case of a male patient presenting with a cholesteatoma of the frontal sinus revealed by neurological complications in order to study the clinical characteristics as well as the treatment and the evolution of this pathology.

Observation

We report the case of a 25-year-old patient with no history of sinus infection or cranial trauma who presented with right painful fronto-orbital tumefaction associated with frontal cephalalgia and a 1-week significant ipsilateral eyelid oedema plus a fever of 40 °C without rhinorrhea. After his admission to the emergency room, the patient experi-

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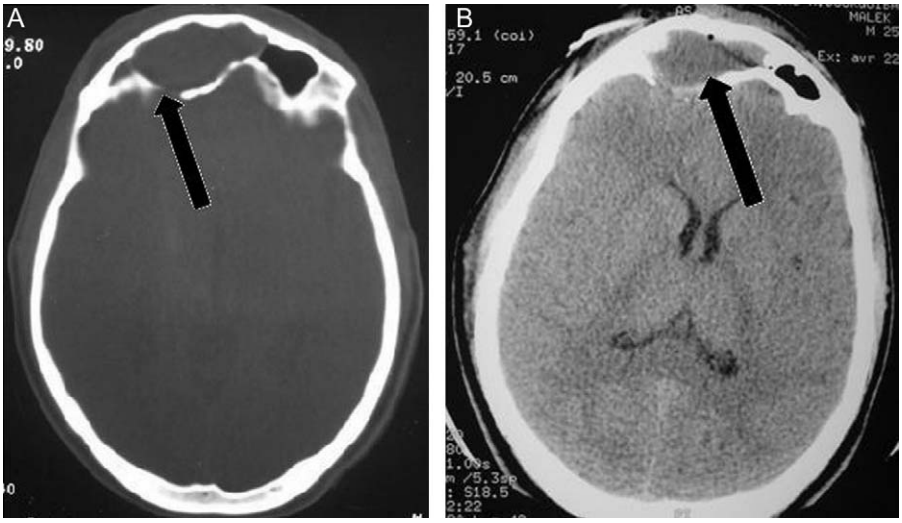

Figure 1 Cranial CT-scan in axial section with iodine contrast injection realized at patient's admission. A. Bone window: obliteration of the frontal sinus with lysis of the right posterior wall (arrow). B. Parenchymal window: obliteration of the right frontal sinus via parenchymatous process (arrow).

enced disorders of consciousness and generalized convulsive seizures that led us to take him to ICU. He required an anticonvulsant treatment and etiological evaluation. During examination, the patient was conscious with a Glasgow score at 8/15. A right 1,5-cm long axis eyelid tumefaction with a significant superior palpebral oedema and a right axial non reductible exophtalmos was observed. Endoscopic examination of the nasal fossa revealed a congestive mucosa without pus in the meatus. Brain and facial bones CT-Scan with iodine injection evidenced complete opacification of the right frontal sinus with bone lysis of the posterior wall without brain alteration (Fig. 1). Eye examination did not reveal any papillary oedema. Complete blood count showed hyperleukocytosis with a predominance of neutrophil polynuclears (PNN) of 28,700 elements per millimetre cube. Erythrocyte sedimentation rate was accelerated at 68 mn at the first hour and C reactive protein was high at 120 mg/l. Renal, hepatic and hemostasis evaluation tests were normal. Lumbar puncture showed a suspicious fluid. Bacteriological analysis of cerebrospinal fluid evidenced Gram-positive cocci with many impaired polynuclears. Culture isolated a *Streptococcus pneumoniae* with a sensitivity reduced to penicillin. The diagnosis of meningocephalitis as an aggravating factor of frontal sinusitis was established. Antibiotic therapy, by general administration with cefotaxime 200 mg/kg per day four times daily, was initiated. Emergency surgery under general anesthesia was realized. The frontal sinus exploration via eyebrow incision after bone flap removal revealed a cyst with secondary infection containing keratin material. Complete surgical resection of the cyst as well as of its shell were realized with careful cleansing of sinus cavities with Betadine® diluted with physiological saline. There was a bone lysis of the posterior wall of the frontal sinus exposing the meninges without leaking of cephalorachidian fluid and a complete stenosis of the nasofrontal canal. Bone flap reinsertion was realized with percutaneous drainage for 3 days. Final histological examination established the diagnosis of cholesteatoma of the frontal sinus. Bacteriological examination of sinus pus isolated a *pneumoniae streptococcus* with a sensitivity reduced to penicillin. Cefotaxime 200 mg/kg per day was administered intravenously for 3 weeks four times a day, then levofloxacine per os 500 mg/day in one take for 3 weeks in the absence of recurrence. Craniofacial MRI with gadolinium injection realized 2 months postoperatively did not evidence any recurrence with a good pneumatization of sinus (Fig. 2). Filling the sinus with autogenous fat material was realized 4 months after the first surgical procedure due to complete stenosis of the nasofrontal canal and lysis of the posterior wall of the frontal sinus. Immediate postoperative period was simple and clinical examination of the patient was normal after a 5-month follow-up.

Discussion

Paranasal sinuses cholesteatoma is a rare occurrence. It can involve all sinuses, most frequently, the frontal sinus and more rarely, the ethmoidal cells and the maxillary sinus [1].

Mean age for cholesteatoma of the frontal sinus occurrence is 48, age ranging from 23 to 80 years [2]. Therefore, our case is among the youngest ones in literature. Two forms are described: the congenital and the acquired form. In the congenital forms, cholesteatoma may be due to the aberrant presence of ectodermal cells during face formation at the 3rd and 5th week of embryonal live [1]. In the acquired forms, three theories were proposed: the first one suggests the existence of iatrogenic implantation of epidermal cells after frontal sinus surgery or trauma [7]. A notion of sinus surgery or cranial trauma has been reported in four out 13 cases in English-speaking literature [2]. The second theory is the theory of epithelial cells migration from nasal vestibule similar to middle ear cholesteatoma but such a migration was never revealed [5]. The third hypothesis suggests the existence of epithelial metaplasia due to chronic rhinosinusitis [7]. Our patient did not experience trauma or chronic rhinosinusitis that could support one of these theories. The clinical signs of frontal sinus cholesteatoma reported in the literature mostly include

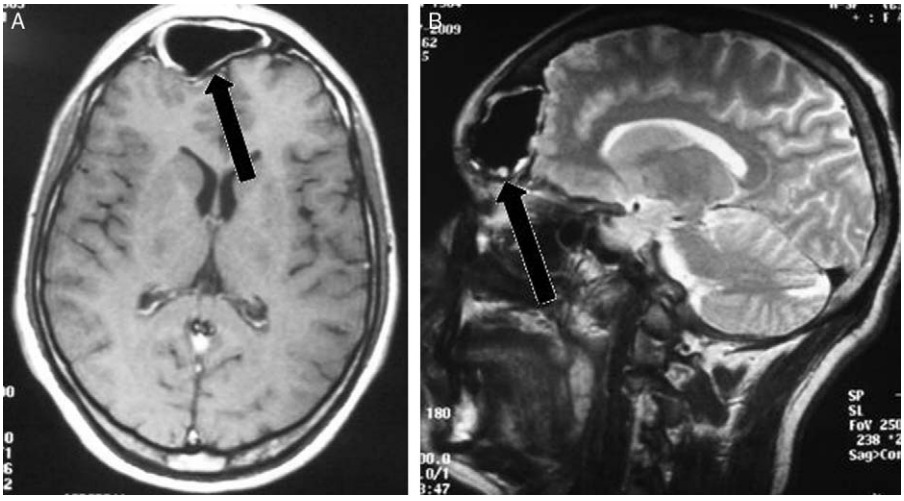

Figure 2 Cranial MRI after 2 months postoperatively showing good pneumatization of the frontal sinus without recurrence (arrow). A. T1-weighted imaging sequence after intravenous contrast in axial section. B. T2-weighted imaging sequence in sagittal section.

frontal cephalalgia fronto-orbital tumefaction with progressive enlargement, ptosis, diplopia or decreased visual acuity [2]. Our patient presented with cholesteatoma with secondary infection associated with bacterial meningoencephalitis. To our knowledge, it was the only case in the literature describing such an evolution of sinus frontal cholesteatoma. Moreover, it was the second reported case of cholesteatoma revealed by an epileptic seizure followed by coma requiring the patient's admission to an intensive care unit [8].

Although not pathognomonic, CT-scan imaging can reveal opacification of the frontal sinus and bone lysis. MRI allows to differenciate purulent sinusitis from tissular lesion [2]. Diffusion sequence reveals the tumoral nature of the cyst, whether it is infected or not.

Standard treatment is complete surgical resection of the cholesteatoma and its shell with careful cleansing of sinus cavity [1,2,5]. The procedure is realized via external approach in order to maintain a better monitoring of sinus cavity [1,2]. Bicoronal Cairns-Unterberger approach was used to provide a better access to sinus area and obtain a concealed scar. Eyebrow incision was a therapeutic alternative. In our case, it allowed us to make a wide enough opening of the frontal sinus permitting the control of the entire sinus area; thanks to the 30°C rigid optics, it was possible to check the cavity and examine the nasofrontal canal.

Complete surgical resection prevents the risk of recurrence all the more so as cases of carcinous degeneration of sinus cholesteatoma were described [3]. Obliteration of frontal sinus is recommended by some authors when the cholesteatoma is associated with sinusitis, in cases of complete stenosis of the nasofrontal canal or when excision cavity is too wide [5]. In our case, we decided to realize an obliteration of the frontal sinus since there was a stenosis of the nasofrontal canal and a lysis of the posterior wall, which caused an almost fatal meningocephalitis for the patient.

The obliteration was realized in second stage because of the sinus infection. This procedure enabled us to check the sinus again and remove the entire inflammatory mucosa.

In case of lysis of the anterior wall due to erosive process, some authors recommend to realize a reconstruction of bone defect 1 year after the first surgery in order to avoid the risk of recurrence [2,5]. As in middle ear cholesteatoma, postoperative follow-up is based on clinical examination (endoscopy of nasal cavities) and especially MRI more accurate and specific than CT-Scan in the detection of cholesteatoma residual lesions or recurrence. Two MRI sequences allow to differentiate cholesteatoma from an inflammatory or scar tissue on delayed image after contrast media injection and diffusion sequences. Contrary to scar tissue, there is no enhancement of cholesteatoma after contrast media injection even on delayed images. On T1 and T2-weighted imaging sequences, cholesteatoma is clearly hyperintense [2].

Conclusion

Frontal sinus cholesteatoma is a rare entity. This pathology must be evoked when a slowly evolutive process associated with bone lysis is observed. Bone destruction can cause complications that can sometimes put the patient's life prognosis at stake. Preoperative diagnosis is difficult to establish because of the lack of specificity of clinical signs and imaging data. Complete removal of the cholesteatoma and its matrix is required to avoid recurrences. Standard follow-up is based on MRI examination.

Conflicts of interest statement

None.

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